

Subtotal capsulectomy for idiopathic chondrolysis of the hip

A clinical, radiological and histological study

by

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PART A:

RESEARCH PROPOSAL

RESEARCH PROPOSAL:

Subtotal capsulectomy for idiopathic chondrolysis of the hip

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Purpose

The purpose of this study will be to review the outcome of a subtotal capsulectomy of the hip for idiopathic chondrolysis. We plan to review the imaging (radiographs, CT scans and MRI scans), histology and clinical outcome. This will be measured against international literature.

Background

Idiopathic chondrolysis of the hip is a very rare condition. It is characterized by cartilage necrosis of the hip joint not associated with trauma, SUFE, infections or other demonstrable causes. It was first described in 1971 by Jones from the Princess Alice Orthopaedic Hospital in Cape Town.

It occurs mainly in adolescent girls. The outcome in South Africa has been reported as a progressive downhill course resulting in a painful, stiff hip. The aetiology of the disease remains unknown. Theories suggested are mechanical (decreased movement with loss of synovial nutrition; increased joint pressure) and an auto-immune response in genetically predisposed individuals.

The differential diagnosis includes atrophic-type tuberculosis of the hip.

Suggested treatment ranges from NSAIDs and range of motion exercises alone to early aggressive surgical treatment. Our experience with continuous passive motion (CPM) and NSAID treatment have been disappointing.

In 1988 Roy and Crawford described a subtotal capsulectomy of the affected hip with concomitant muscle releases to relieve intra-articular pressure and correct muscle contractures.

This treatment was tried at our unit with encouraging results. In 2011 a treatment protocol was started that involved:

- Pre-op
 - Routine blood work-up and Mantoux skintest to rule out TB
 - Radiographs, CT scan and MRI scan of the affected hip for diagnosis
 - Trial of non-operative management: NSAID's, traction and CPM
- Subtotal capsulectomy with muscle releases if no response to non-operative management
 - Histology and culture of synovium and cartilage to confirm diagnosis
- Post-op
 - NSAID's and CPM
 - Follow up radiographs and MRI scan to assess disease progression

Subsequently we've performed 5 such cases with biopsy of the affected cartilage at the same time. We want to assess the outcome of this treatment.

Materials and Methods

Study design

This will be a retrospective cohort study. We will perform a retrospective review of prospectively collected data on these patients.

Study population

Inclusion criteria

All patients that had a subtotal capsulectomy of the hip performed for idiopathic chondrolysis from January 2011 to August 2012.

Exclusion criteria

All patients with less than 6 months follow up after surgery.

All patients with inadequate follow up notes.

Recruitment

The theatre records at Maitland Cottage Hospital will be used to identify the patients from January 2011 to August 2012.

Data collection

The patients' hospital folders will be accessed and the orthopaedic notes will be used to collect the relevant clinical data. Laboratory results will be collected from the electronic National Health Laboratory System. Images will be reviewed on the electronic Picture Archiving and Communication System (PACS).

The following data will be collected from the folders

- Demographic information: Age, gender
- Presenting symptoms and duration
- Clinical findings at presentation, specifically pain and range of motion of the hip
- Results of laboratory tests
- Findings of imaging, including radiographs, computed tomography (CT) scans and magnetic resonance imaging (MRI) scans
- Details of surgery performed
- Results of histology of cartilage biopsy
- Clinical findings at final follow up, specifically pain and range of motion of the hip
- Findings of follow up imaging

Data analysis

After collection Microsoft Excel tables and graphs will be used to for data capturing and analysis.

Risks and benefits

There will be no risk to the participants as this is a retrospective review of investigations and procedures already performed. No additional investigations or procedures will be performed.

Benefits would include measuring the outcome of procedures performed in our unit and being able to compare this to international literature. We would be able to review our practise and better inform future patients of the expectant outcome of the surgery. We would also be able to contribute to the current body of literature on the subject.

Informed consent

No consent will be obtained as this is a retrospective review of cases already performed.

Privacy and confidentiality

Privacy and confidentiality of all data and results will be ensured. All data is delinked once we have completed data collection, but obviously as the treating surgeon we will recognise the pathology so anonymity is limited. We never put names in our presentations or publications.

Outcome of study

Our aim would be to publish our results in a paediatric orthopaedic journal for peer review. We also aim to present our results at the annual South African Orthopaedic Congress.

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PART B:

LITERATURE REVIEW

LITERATURE REVIEW:

Subtotal capsulectomy for idiopathic chondrolysis of the hip

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Introduction

Idiopathic chondrolysis (IC) of the hip is a very rare, crippling condition affecting mainly adolescent females with fewer than 130 cases documented.¹ It is characterized by rapid cartilage necrosis of the hip joint, not associated with trauma, slipped upper femoral epiphysis (SUFE), infection or other demonstrable causes.² Patients present with insidious onset of pain, a limp and a typical flexion, abduction and external rotation deformity of their hip joint and radiological loss of joint space of the affected hip.

The natural history ranges from complete recovery to painful ankylosis.¹ Management remains controversial and various treatment strategies have been advocated. Non-operative treatment with non-steroidal anti-inflammatory drugs (NSAIDs), non-weightbearing and physiotherapy, including continuous passive motion (CPM), are used.³⁻⁵ Aggressive operative treatment has also been described. In 1988 Roy and Crawford reported on a subtotal capsulectomy with soft tissue releases followed by aggressive rehabilitation.⁶ Three cases were treated with good results. Subsequent authors though reported indifferent results with the same technique.⁷

Objectives

The objectives of this literature review are to:

- Clarify the definition, epidemiology, aetiology, patho-anatomy, clinical and radiographic features and natural history
- To research the treatment options used and critically analyse the outcomes of these described methods
- To identify areas of potential future research

Methods

Google Scholar and Pubmed internet search engines were used to search online databases. Review articles on the subject were analysed and their references were researched. The section on idiopathic chondrolysis in the chapter on other conditions of the hip in Lovell & Winter's Pediatric Orthopaedics, 6th Edition² was also reviewed and the relevant references researched. All available evidence was screened and the relevant articles were obtained from the University of Cape Town library.

When searching 'idiopathic chondrolysis of the hip' on Google Scholar 951 articles are cited. The same search on Pubmed cites 38 articles. Similar searches for 'idiopathic chondrolysis managed by subtotal capsulectomy' yielded 22 and 1 articles respectively.

Quality of evidence

The evidence on the topic is limited to case studies and small case series. All the studies can be classified as levels 4, 5 or 6 of evidence and there is no higher level of evidence available on the topic.

Summary of literature

Background

Waldenström first described chondrolysis of the hip joint.⁸ It has been described secondary to a variety of conditions, including SUFE, trauma, septic arthritis, severe burns, prolonged immobilization and even Marfan and Stickler syndromes.¹ The term idiopathic chondrolysis refers to chondrolysis where a demonstrable cause cannot be identified.

IC was first described in 1971 by Jones from the Princess Alice Orthopaedic Hospital in Cape Town. He reported on a series of 9 adolescents (8 girls and 1 boy) with hip joint disease that did not conform to other known conditions and that presented with consistent enough clinical pictures to justify the recognition of a distinct clinical entity.⁹ In 1982 Sparks and Dall from the same unit reported on the original series and a further 9 patients. They confirmed the existence of the condition and reported a progressive downhill course with a poor outcome in 88% of cases despite different treatment strategies.¹⁰ They also confirmed that there were no subsequent developments of systemic illness with longer follow up of the original group. Duncan in 1975 coined the term

'idiopathic chondrolysis' to differentiate the condition from chondrolysis secondary to other causes.¹¹ Subsequently the condition was verified with reported cases from around the world.¹¹⁻²¹

Definition

The presence of stiffness accompanied by the radiographic hallmark of osteopenia and concentric reduction of the joint space to less than 3mm can be considered a diagnostic criterion for IC.^{1,2}

Epidemiology

The true incidence of IC is unknown. Fewer than 130 cases have been documented in the literature.¹ Some authors believe that it might be a more common cause of degenerative arthritis in woman.¹⁸ IC affects mostly a unilateral hip, but bilateral cases have been described.^{22,23} It affects mainly adolescents with a mean age of onset of 11 quoted in the literature¹, although cases presenting shortly after skeletal maturity have been described.²⁴ Girls are affected six times more than boys.² Initially it was thought to occur in individuals of African descent only, but subsequently cases have been documented from different races across the world.^{2,24}

Chondrolysis has been described in various other joints, including the knee, ankle, shoulder and elbow.^{25,26} Chondrolysis following arthroscopy were reported in both the knee and shoulder.²⁵

Aetiology

The aetiology of the condition remains unknown. Various theories have been suggested, including auto-immune and mechanical theories. Some authors even suggest IC to be a form of cartilaginous dysplasia.²⁷

Auto-immune theories

The most widely accepted theory is that the articular cartilage destruction is due to an auto-immune response in genetically susceptible individuals.^{1,2,28} Supporters of this theory have shown inflammatory cell infiltrates and immunoglobulin deposits in the synovium of the hip joints.^{12,21,29-31} Some authors have shown serological abnormalities.² Ippolito et al noted an absence of the chronic degenerative changes seen in osteoarthritis.²⁹ They concluded that idiopathic chondrolysis is a pathological process which develops and runs its course very quickly.

Mechanical theories

Others have proposed various mechanical theories, including a decrease in synovial nutrition,^{8,32} a mechanical insult resulting in release of chondrolytic enzymes² or an increase in intracapsular pressure.⁶ In SUFE the mechanical theory is supported by the fact that persistent pin or screw penetration only in the antero-superior weight-bearing quadrant of the femoral head resulted in chondrolysis.³³ Hips with transient pin or screw penetration or penetration of the postero-inferior quadrant did not develop chondrolysis. Removal of the penetrating metalware had a 75% chance of recovery. Those patients however who had chondrolysis at presentation were all female of African descent with chronic moderate and severe slips, suggesting a genetic and mechanical cause. The results of these hips are poor in the majority of cases,³⁴ which is similar to that of IC reported from the same unit.¹⁰

Patho-anatomy

The femoral side of the hip joint is affected primarily according to the literature, with changes starting in the weight-bearing area and spreading peripherally.¹ Articular cartilage biopsies show thinning of the superficial zone, with chondrocyte necrosis. The basal zone contains some viable chondrocytes with disorganised collagen formation.^{24,29}

Biopsies of the synovium show oedematous, hyperplastic synovium. There are features of a chronic inflammation with a perivascular infiltrate of lymphocytes and plasma cells. Some authors have shown immunoglobulin deposits in the synovium of the hip joints^{12,29-31} although others contradict these findings and report immunofluorescent studies for immune complex deposition to be normal.³⁵ No granuloma formation is seen.²

Clinical features

The typical history is of insidious onset over 2 to 3 months of pain in the groin, anterior thigh or knee with progressive stiffness of the hip joint and a limp.¹ Examination reveals an antalgic, Trendelenburg gait with a typical flexion, abduction and external rotation deformity. There is normally a reduction in movement in all planes and if presentation is delayed patients can present with fixed contractures. Secondary pelvic obliquity, apparent leg length inequality and an increase lumbar lordosis can also occur. There is an absence in systemic symptoms and signs.¹

There appears to be two distinct stages in the disease with an initial acute stage with onset of symptoms caused by an inflammatory response. This is followed by chronic phase with an unpredictable course.^{1,2}

Laboratory features

Laboratory tests are conducted to exclude other conditions as positive findings preclude the diagnosis of IC. Full blood count, including white cells, blood cultures, inflammatory and auto-immune markers and Mantoux skin test should all be within normal limits. C-reactive protein (CRP) serum levels are within normal levels, but a slightly elevated erythrocyte sedimentation rate (ESR) of up to 30 mm/h has been reported in some cases.³⁶ Markers of auto-immune diseases, including rheumatoid factor, antinuclear factor and anti-neutrophil cytoplasmic antibodies are negative. Human leucocyte antigen-B27 surface antigen evaluation is also reported to be normal.³⁵

Imaging

Plain radiography

Plain radiography is the initial investigation of choice and in the appropriate clinical setting, no further radiological investigation is necessary. Although radiographs can be normal early on in the disease process, it is useful to exclude other conditions. Typical early radiographic changes include peri-articular osteopenia, joint space narrowing, small subchondral irregularities and pelvic tilt due to the contractures around the hip joint.³⁵

Later in the disease widening of the femoral neck and epiphysis³⁷ and early closing of the femoral capital physis is seen,¹⁵ as is protrusio.³⁸ Widening of the pubic symphysis has also been described.² Features are similar to osteoarthritis, but with preservation of the sphericity of the femoral head.¹⁰ Some authors have postulated that IC and primary acetabular protrusio (PAP) are due to the same disease process,³⁸ although these are mostly recognised as distinct entities. Protrusio acetabuli is seen in up to 50% of late radiographs.¹

Computed Tomography (CT)

CT, though superior in depicting anatomical detail and in its sensitivity for detecting subchondral bone erosions, its benefit over and above plain radiography is negated by the radiation burden, particularly in the vulnerable pediatric setting.

Magnetic resonance imaging (MRI)

MRI can be used to aid diagnosis where it is unclear. Changes manifest earlier than on plain film.¹ In addition to the standardized anatomical imaging planes using conventional joint imaging protocols, cartilage specific imaging sequences are employed to distinguish the different zones of

cartilage (physeal, epiphyseal and articular cartilage) in children. In addition to the routine sequences used for the evaluation of cartilage in adults (intermediate/ proton density with fat suppression and T1-weighted spoiled gradient recalled echo with fat suppression) T2-weighted sequences that help differentiate the low signal epiphyseal cartilage from the higher signal physeal and articular cartilages are also employed.³⁹

Johnson et al divided MRI findings into early and late features. Early features included focal cartilage loss, marrow oedema in the femoral head and adjacent acetabulum, a small joint effusion without significant synovial enhancement, muscle wasting and signs of bone remodelling.^{35,37,40} Cartilage loss is described as mostly centrally from the femoral head.^{1,37} Late features were more extensive marrow oedema and widespread cartilage loss and bone erosions.³⁷ Slight synovial enhancement has been described in the early phase,³⁵ but significant synovial enhancement is diagnostic of juvenile idiopathic arthritis or chronic infections, like TB.

Other imaging

Bone scintigraphy, when performed, shows normal flow and bone uptake but diffuse peri-articular uptake suggestive of a diffuse inflammatory process.¹⁸

Differential diagnosis

IC is a diagnosis of exclusion and the differential is wide. Septic arthritis of the hip must be excluded. In South Africa where tuberculosis (TB) is prevalent, the main differential is atrophic-type TB of the hip.⁴¹ In TB femoral and acetabular erosions are seen on plain radiographs.⁴²

Chondrolysis secondary to other causes must also be excluded. A variety of other conditions can present in a similar manner, namely SUFE (with or without chondrolysis), Perthes' disease, trauma, PAP, juvenile idiopathic arthritis (although the hip is affected very rarely in the pauciarticular form) and pigmented villonodular synovitis.¹ Some authors even suggest IC and juvenile idiopathic arthritis (JIA) are the same entity and should be treated as such.²¹

Natural history

The natural history of idiopathic chondrolysis is unpredictable. Originally it was thought to follow a relentless progressive course towards malpositioned fibrous ankylosis.¹⁰ Since then a variety of outcomes from spontaneous resolution to painful ankylosis have been described.^{1,9,10} Several authors describe spontaneous resolution of the condition over time.^{15,16,43} Some authors even

describe restoration of the joint space with up to 2mm over time.¹ It is said that an acceptable clinical outcome can be achieved in 50 to 60% of cases with conservative treatment.¹

There appears to be two distinct stages in the disease. An initial acute phase is characterised by an inflammatory response with cartilage destruction and worsening hip pain and decreased hip motion over up to 18 months.¹ This is followed with a chronic phase that is less predictable and lasts three to five years.² There appears to be three distinct clinical outcomes with half the patients showing clinical improvement over time. Even complete resolution is reported in extreme cases. The second group develops a stiff hip that limits function, but is pain free. The third group regresses to a painful, malpositioned ankylosis. Currently there are no known indicators to predict the individual outcome of the chronic phase.¹

Treatment options

Initially early, aggressive operative treatment of IC was practised, including soft tissue releases, corrective osteotomies, arthrodesis and arthroplasty.¹ Better understanding of the natural history led to a more conservative approach. Non-operative treatment aims at relieving pain, maintaining range of motion and offloading the hip joint. There is no general consensus on how this is best achieved. A treatment protocol of NSAIDs, prolonged protected weightbearing, aggressive physiotherapy and periodic traction have shown clinical improvement in 50% to 60% of cases.^{15,16} Continuous or intermittent traction may also improve the condition, although this is needed for up to 3 months and not without associated morbidity.¹ CPM has also been reported to have a positive effect.³ The possible auto-immune cause suggests that the powerful anti-inflammatory properties of the tumour necrosis factor-blockers might be useful. There is a single case study of successful treatment with etanercept.⁴⁴

Operative treatment is reserved for patients who have failed prolonged non-operative treatment. A wide variety of surgical interventions have been described with variable success. Early surgical interventions include soft-tissue releases to correct hip deformity, capsulectomy and articulated hinged distraction. Aldegheri et al reported clinical improvement despite radiological deterioration in cases of articulated distraction for chondrolysis secondary to SUFE.⁴⁵ Articulated distraction has also been described for the adolescent arthritic hip due to other causes with encouraging results.⁴⁶ Salvage procedures include excision arthroplasty, arthroplasty and arthrodesis. Some authors suggest that arthrodesis is the gold standard salvage procedure in IC.²

Subtotal capsulectomy with soft tissue releases

Roy and Crawford⁶ in 1988 described a subtotal capsulectomy with soft tissue releases and aggressive rehabilitation in three cases. They performed a subtotal (270°) circumferential capsulectomy excising a 1 cm strip of capsule, sparing the posterior 90° to preserve blood supply to the femoral head, while taking care not to injure the labrum. Concomitant soft-tissue releases were performed as necessary to address joint contractures. The origin of the sartorius muscle and the straight head of the rectus femoris muscle were released. A psoas tenotomy was performed at the pelvic brim and the tensor fascia lata was released proximally to improve the abduction contracture. This was followed by aggressive rehabilitation, including the use of CPM.⁶ Although intra-articular pressure was never measured, they postulated that an increase in intra-articular pressure was relieved. One case received another manipulation under anaesthesia. They described good results at an average of more than 3 year follow up with all patients being symptom free with improvement in range of motion. They also reported restoration of the joint space on radiographs in all cases and therefor justified aggressive early surgical intervention.⁶ Laor and Crawford³⁵ later reported on a series of seven hips in six patients that all underwent the same procedure. Only three cases showed clinical and radiological resolution following the procedure with another showing moderate joint space narrowing. The remaining three hips developed severe ongoing loss of joint space resulting in total hip replacements in two hips at a young age.

Other authors reported clinical and radiological progression of the disease following similar surgical procedures³⁸ and early clinical improvement with radiological deterioration and no long term benefit.⁷ Korula et al⁴⁷ reported on the largest series of surgically managed IC cases with an average follow up of 23.4 months. They performed a similar subtotal capsulectomy and found a small, but statistically significant improvement in hip range of motion. The pre-operative arc was improved slightly and was brought into a more functional range postoperatively.

The current recommendations from the literature are to control inflammation and ROM in the acute stage. This is achieved through the use of NSAIDs, physiotherapy, protected weightbearing, CPM and traction. Surgical intervention is reserved for cases where prolonged non-operative treatment fails.

Areas for further research

Idiopathic chondrolysis remains a challenge to physicians in both the aetiology and treatment. Basic science research can hopefully shed the light on the exact aetiology and pathogenesis of the cartilage degradation.

Various alternative treatment modalities should be further researched. As previously mentioned the possible auto-immune cause suggests that the powerful anti-inflammatory properties of the tumour necrosis factor-blockers might be useful. There is a single case study of successful treatment with etanercept and this needs to be further researched.⁴⁴ Salter in 1980 demonstrated the effect of CPM on healing of full thickness defects in articular cartilage.⁴⁸ In theory it should be possible to address acetabular cartilage loss with chondrogenesis through metaplasia by drilling subchondral bone and CPM.

The rarity of the condition makes research difficult as even large referral centres might only see up to one case per year.

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PART C:

MANUSCRIPT

This manuscript will be submitted to the Journal of Pediatric Orthopaedics. The format and referencing style is according to the instructions for the authors of the journal (see attached instructions in Part D).

ARTICLE:

Subtotal capsulectomy for idiopathic chondrolysis of the hip

A clinical, radiological and histological study

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Abstract

Background

Idiopathic chondrolysis (IC) of the hip is a rare, crippling condition affecting mainly adolescent females. Cartilage necrosis results in a stiff, painful hip which in the majority of cases have a poor outcome. The aetiology still remains unknown and the treatment controversial and relatively unsuccessful. Subtotal capsulectomy with soft tissue releases has been reported to give good results.

Methods

Five female adolescents with IC of the hip were reviewed in order to shed light on the aetiology, delineate the role of magnetic resonance imaging (MRI) pre- and post-operatively and assess the outcome of a subtotal capsulectomy. The surgical technique was performed as originally described with concomitant releases of the deformities. Samples of synovium and cartilage were sent for histology and culture (including tuberculosis culture). Pre- and post-operatively the patients were evaluated regarding pain, deformity and range of motion (ROM). Follow up radiographs and MRI scans were performed and compared to pre-operative imaging.

Results

Blood tests for auto-immune markers were negative in all cases. MRI pre-operatively showed erosions and mainly destruction of the superomedial acetabular cartilage. Histology of the synovium showed chronic, non-specific inflammation with a plasma cell infiltrate, suggesting an auto-immune cause. Histology of the cartilage confirmed cartilage necrosis. At follow up (mean 11 months) four of the five patients were pain free, but all five had an improvement in pain. There was an improvement in deformity and range of motion in four cases. Post-operative imaging (radiographs and MRI) at a mean of eight months, however, showed deterioration of the pathology with increase of the erosions, joint space narrowing and cartilage destruction.

Conclusions

We conclude that the aetiology is most likely an auto-immune process. MRI was helpful in delineating the cartilage pathology. A subtotal capsulectomy offers early post-operative clinical relief. Radiological deterioration however suggests that the surgery does not prevent deterioration of pathology.

Level of Evidence

Level IV: Case series

Key words: Idiopathic chondrolysis, hip, subtotal capsulectomy

Introduction

Idiopathic chondrolysis of the hip is a very rare disorder that occurs mostly in adolescents with fewer than 130 cases documented¹. It is characterized by rapid cartilage necrosis of the hip joint, not associated with trauma, slipped upper femoral epiphysis (SUFE), infection or other demonstrable causes.² Patients present with insidious onset of pain, a limp and a typical flexion, abduction and external rotation deformity of the hip joint.

It was first described in 9 adolescents by Jones in 1971. The disease did not conform to other known conditions justifying the recognition of a distinct clinical entity.³ In 1982 Sparks and Dall from the same unit reported on the original series and a further 9 patients. They confirmed the existence of the condition and reported a progressive downhill course with a poor outcome in 88% of cases despite different treatment strategies.⁴ Subsequently the condition was verified with reported cases from around the world.⁵⁻¹⁵

The aetiology remains unknown. Theories include an auto-immune response in genetically susceptible individuals^{5,16} and mechanical theories such as a decrease in synovial nutrition¹⁷, a mechanical insult with release of chondrolytic enzymes² and an increase in intracapsular pressure.¹⁸

The presence of stiffness accompanied by radiographic osteopenia and concentric reduction of the joint space to less than 3mm can be considered a diagnostic criterion for IC.^{1,2} In our setting the main differential diagnosis is atrophic-type tuberculosis of the hip.^{19,20}

The natural history of idiopathic chondrolysis is unpredictable. A variety of outcomes from spontaneous resolution to painful ankylosis have been described.^{1,3,4} There appears to be two stages in the disease with an initial acute stage with onset of symptoms caused by an inflammatory response. This is followed by a chronic phase with an unpredictable course.^{1,2}

Various treatment strategies have been advocated. Non-operative treatment with non-steroidal anti-inflammatory drugs (NSAIDs), non-weight bearing and physiotherapy, including continuous passive motion (CPM), are used.²¹⁻²³ In 1988 Roy and Crawford reported on a subtotal capsulectomy with soft tissue releases followed by aggressive rehabilitation.¹⁸ Three cases were treated with good results. Subsequent studies reported indifferent results with the same technique.²⁴

The purpose of our study was to try and shed light on the aetiology, to delineate the role of MRI and to assess the outcome of subtotal capsulectomy.

Materials and Methods

Five adolescent female patients with a mean age of 11 years (range 10 to 12 years) treated from January 2011 to August 2012 were reviewed. During this period no patient was treated successfully non-operatively. The median duration of symptoms was 2 months (range 2-24 months). We performed a retrospective review of prospectively collected data on these patients. No patients were excluded from analysis.

Pre-operatively the patients were assessed clinically. Blood work-up included full blood count, erythrocyte sedimentation rate (ESR), rheumatoid factor, antinuclear factor and anti-neutrophil cytoplasmic antibodies. A chest radiograph and a Mantoux skin test were done. Pelvic radiograph, computed tomography (CT) and MRI scans of the hip were done. In addition to the standardized MRI anatomical imaging planes using conventional joint imaging protocols, cartilage specific imaging sequences were employed to distinguish the different zones of cartilage (physeal, epiphyseal and articular cartilage) in children. In addition to the routine sequences used for the evaluation of cartilage in adults (intermediate / proton density with fat suppression and T1-weighted spoiled gradient recalled echo with fat suppression) we also employed T2-weighted sequences that help differentiate the low signal epiphyseal cartilage from the higher signal physeal and articular cartilages.²⁵

All patients underwent an extensive course of non-operative management that included NSAIDs, physiotherapy and CPM. All five patients underwent subtotal capsulectomy with soft tissue releases. The surgical technique was as described by Roy and Crawford.¹⁸ A 1 cm strip of hip capsule was excised for an arc of 270°. The posterior 90° was spared to preserve the blood supply to the femoral head. A psoas tenotomy at the pelvic brim and a release of the straight head of rectus femoris were done to release the flexion contracture of the hip. The tensor fascia lata was released to improve the abduction deformity. Synovial biopsies were sent for histology and culture (including TB culture) and a cartilage biopsy taken from the periphery of the femoral head was sent for histology.

Postoperatively the patients were initially treated in bilateral skin traction. After 7 to 10 days physiotherapy, including CPM was initiated. Patients received NSAID's for 6 weeks.

At a mean follow up of 11 months (range 9 to 16 months) the patients were assessed for pain and ROM. Follow up radiographs and MRI scans were done at a mean of 8 months to assess the disease progression.

Results

All the patients presented with pain and stiffness of the affected hip. Examination revealed an antalgic, Trendelenburg gait and a painful, stiff hip with a flexion, abduction and external rotation deformity in all cases (Table I). All blood tests including the auto-immune markers were normal. Chest radiographs were clear and Mantoux skin tests negative in all cases.

Initial pelvic radiographs revealed osteopenia, concentric joint space narrowing (less than 3mm) and the deformity was visible in all cases (Figure 1). In one case subchondral erosions were visible on plain radiograph. Two cases fulfilled radiological criteria for protrusio bilaterally. CT confirmed osteopenia and joint space narrowing in all cases, and subchondral erosions were seen in three (Figure 2). MRI scan at presentation was performed in four patients (it was not performed in one case due to non-availability of the scanner). In all four cases where an initial MRI was performed joint space narrowing and marrow oedema was seen (Figure 3-A). Subchondral erosions were seen in three out of four cases (Figure 3-B). The cartilage loss and erosions were mostly acetabular and mainly in the superomedial weight bearing area of the acetabulum (Figure 3-D). Slight synovial enhancement was seen in one case.

Non-operative treatment failed in all cases. Histological examination of the synovium showed signs of mild, non-specific chronic inflammation with a perivascular infiltrate of lymphocytes and plasma cells in all cases. There was no evidence of granulomatous inflammation (Figure 4-A).

Macroscopically, the cartilage appeared lustreless. Microscopically, there was an outer layer of fibroblastic tissue with an adjacent layer of degenerate chondrocytes. There were viable chondrocytes at the base (Figure 4-B). Microbiological culture was negative in all cases, including extended TB culture.

At last follow up all patients subjectively felt that their pain improved. Four patients felt that their hips were pain free. The gait improved in four out of the five cases. The deformity was improved in four cases. In one case the patient's hip was still painful and there was no improvement in the deformity (Table I).

Post-operative radiographs showed deterioration of the joint space narrowing in four of the five cases. On MRI subchondral erosions were now visible in all five hips and cartilage destruction was worse compared to the pre-operative MRI in all four cases with pre-operative MRI.

Discussion

Idiopathic chondrolysis occurs mostly unilateral, although bilateral cases have been described.^{26,27} It affects adolescents with a predominance among girls, but male cases and cases presenting shortly after skeletal maturity have been described.²⁸ Initially it was thought to occur in individuals of African descent only, but subsequently cases have been documented from different races across the world.^{2,28} Literature is limited to case studies and small case series.

Aetiology

The most widely accepted theory is that the articular cartilage destruction is due to an auto-immune response in genetically susceptible individuals.^{1,2,29} Supporters of this theory have shown inflammatory cell infiltrates and immunoglobulin deposits in the synovium of the hip joints.^{5,16,30,31} Some authors have shown serological abnormalities.² In our series auto-immune markers were all negative. A slightly elevated ESR of less than 30mm/h has been reported.³² The synovial inflammatory infiltrate, especially plasma cells, supports the theory of an auto-immune disease. This suggests that the powerful anti-inflammatory properties of the tumour necrosis factor-blockers might be useful. There is a single case study of successful treatment with etanercept.³³

Mechanical theories are a decrease in synovial nutrition,^{17,34} a mechanical insult with release of chondrolytic enzymes² or an increase in intracapsular pressure which is supposedly released by subtotal capsulectomy.¹⁸ In SUFE the mechanical theory is supported by the fact that persistent pin or screw penetration in the antero-superior weight-bearing quadrant of the femoral head resulted in chondrolysis.^{35,36} Hips with transient penetration or penetration of the postero-inferior quadrant did not develop chondrolysis. Removal of the penetrating metal ware had a 75% chance of recovery. Those patients however, who had chondrolysis at presentation before surgery, were all female of

African descent with chronic moderate or severe slips, suggesting a genetic and mechanical cause. The results of these hips are poor in the majority of cases,³⁶ which is similar to that of IC reported from the same unit.⁴

Some authors suggest IC to be a form of cartilaginous dysplasia.³⁷

Ippolito et al in an ultrastructural study of the articular cartilage in IC found no resemblance to the chronic degenerative changes seen in osteoarthritis. They concluded IC to be a pathological process which, unlike osteoarthritis, runs its course very quickly.³⁰

Imaging

Plain radiography is the initial investigation of choice and in the appropriate clinical setting, no further radiological investigation is necessary. Radiographs are frequently normal early on in the disease process.¹ Subsequent findings reveal joint space narrowing, juxta-articular osteopenia and small subchondral irregularities together with remodelling of the femoral neck and epiphysis.³⁸ Premature closure of the femoral capital physis⁹ and protrusio acetabuli³⁹ is also documented. Late manifestations of the disease reflect the secondary degenerative changes of osteoarthritis.⁴

Our investigation demonstrated the typical plain radiographic findings of joint space narrowing, juxta-articular osteopenia and joint deformity. In addition, two cases revealed bilateral protrusio acetabuli according to radiographic criteria. Some authors have implicated the same disease process in IC and primary acetabular protrusio despite their distinct natures.³⁹ Only one case in our series demonstrated subchondral erosions on the initial radiograph.

CT, though superior in depicting anatomical detail and in its sensitivity for detecting subchondral bone erosions (three out of five cases at presentation), its benefit over and above plain radiography is negated by the radiation burden, particularly in the vulnerable pediatric setting. Hence this has subsequently been excluded from our diagnostic workup.

We found MRI useful in aiding diagnosis in difficult cases as well as assessing progression of the disease. MRI findings are divided into early and late features. Early features included focal cartilage loss, bone marrow oedema, a small joint effusion without significant synovial enhancement, muscle wasting and signs of bone remodelling.^{38,40,41} Late features were widespread cartilage loss and bone erosions.³⁸

Our case series demonstrated similar features with the notable exception of early articular cartilage destruction afflicting the acetabular cartilage (superomedial weight-bearing areas) more than the femoral head cartilage as suggested by earlier reports.^{38,40} Synovial enhancement was documented in one of four of the initial MRI studies in our series. Slight synovial enhancement has been described in the early phase of IC.⁴⁰ Significant synovial enhancement is however more suggestive of juvenile idiopathic arthritis (JIA) or chronic infections including TB. MRI is therefore useful in both the diagnostic workup and for the follow-up of disease progression in IC and is routinely employed in suspected cases in our practice.

The only realistic differential diagnosis to be entertained in the local clinical scenario is that of atrophic-type tuberculosis of the hip. A study by Reardon et al compared radiographs of 8 patients with IC and 9 with atrophic tuberculosis. Plain radiographs of the TB hips showed peri-articular erosions which were absent in IC.²⁰ In the current study three hips had subchondral erosions at presentation on CT and MRI, mainly superomedially in the acetabulum. Monoarthritis of the hip in JIA, is invoked as a likely mimic in the literature¹, but has never been documented at our institution.

Outcome

Several authors describe spontaneous resolution of the condition over time.^{9,10,42} This is not our experience, with none of our patients responding to prolonged non-operative management.

An interesting finding was that the ROM improved under general anaesthetic prior to surgery in all cases. This confirms that muscle spasm contributes to the deformity as shown by previous authors.³

Roy and Crawford in 1988 described a subtotal capsulectomy with soft tissue releases and aggressive rehabilitation in three cases.¹⁸ Although intracapsular pressure was never measured they felt it relieved increased intracapsular pressure. They described good results with improvement in pain and range of motion in all cases, which was reproduced by some authors.¹³ They also reported restoration of the joint space on radiographs.¹⁸ Other authors however reported clinical and radiological progression of the disease following similar surgical procedures^{39,40,43} and early clinical improvement with radiological deterioration and no long term benefit.²⁴ In all our cases the pain was improved following surgery, with four out of five cases pain free. The deformity was also improved in four cases, with a slight improvement of the ROM of the hip. In all five cases though there was radiological progression. On plain radiograph there was deterioration of the joint space

narrowing and more subchondral erosions were visible. Follow up MRI revealed more severe cartilage destruction in all cases. From these findings we conclude that although the subtotal capsulectomy offers some relief of pain and deformity, it does not alter the progression of the disease. It does however allow the hip to stiffen up in a more favourable position.

Other treatment modalities have been described. Aldegheri et al reported clinical improvement despite radiological deterioration in cases of articulated distraction for chondrolysis secondary to SUFE.⁴⁴ Articulated distraction has also been described for the adolescent arthritic hip due to other causes with encouraging results.⁴⁵ Our experience with this technique in chondrolysis in SUFE was disappointing. Salter et al in 1980 demonstrated the effect of CPM on healing of full thickness defects in articular cartilage.⁴⁶ In theory it should be possible to address acetabular cartilage loss with chondrogenesis through metaplasia by drilling subchondral bone and CPM. The viable chondrocytes shown in the basal cartilage at histology may benefit from this treatment. The use of biological agents like etanercept is also a possibility as discussed earlier.³³ More research is needed on these novel treatments.

Limitations

The main limitation of our study is the retrospective nature as well as the lack of a clinical scoring system to evaluate outcome. We also had a small series, but due to the extreme rarity of the condition most studies on the topic are limited to small case series.

Conclusions

We conclude that the aetiology is most likely an auto-immune process. MRI was helpful in delineating the cartilage pathology, which was mainly on the acetabular side. A subtotal capsulectomy offers early post-operative clinical relief. Radiological deterioration suggests that the surgery does not prevent progression of the pathology.

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Tables

Table 1: Pre- and latest follow-up deformity and ROM

	Mean value (range)	
	Pre-operative	Follow-up
Deformity		
Fixed-flexion deformity	31° (20°-45°)	26° (20°-30°)
Abduction deformity	22° (10°-30°)	13° (0°-30°)
External rotation deformity	20° (10°-30°)	8° (0°-30°)
Range of motion		
Flexion-extension arc	25° (0°-60°)	40° (30°-60°)

Figures

Figure 1: A-P radiograph of the pelvis of a 11-year-old girl with IC of the right hip showing typical findings of osteopenia, joint space narrowing (JSN), closure of the femoral capital physis and flexion, abduction and external rotation deformities.



Figure 2: Coronal CT scan of the same patient confirming joint space narrowing and also showing subchondral erosions.

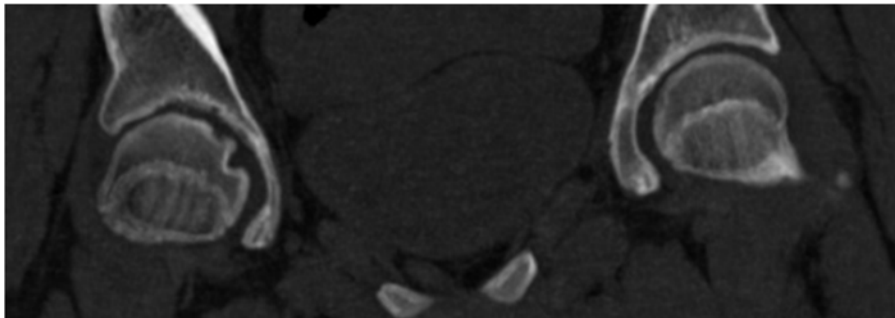
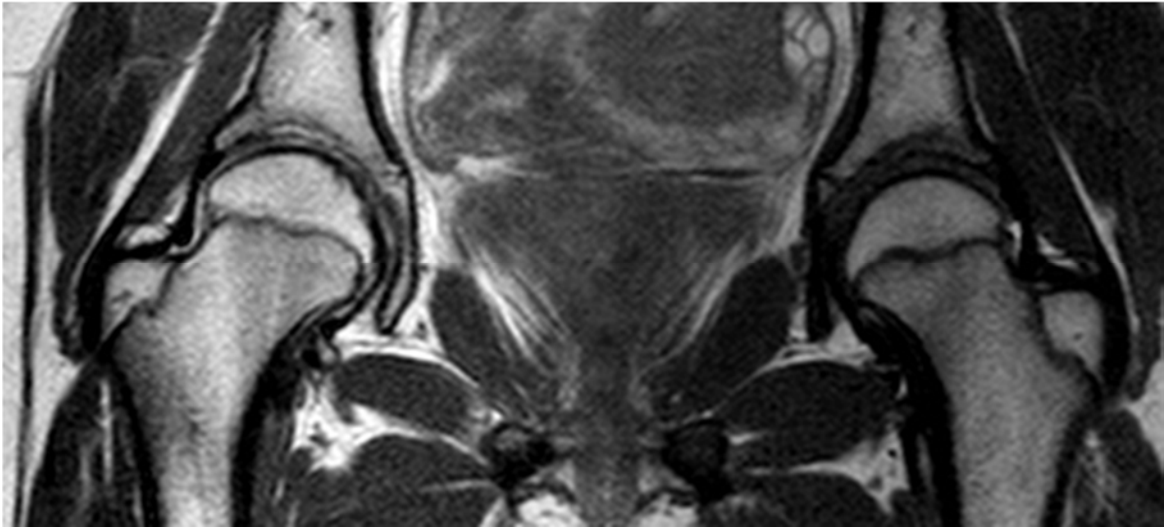
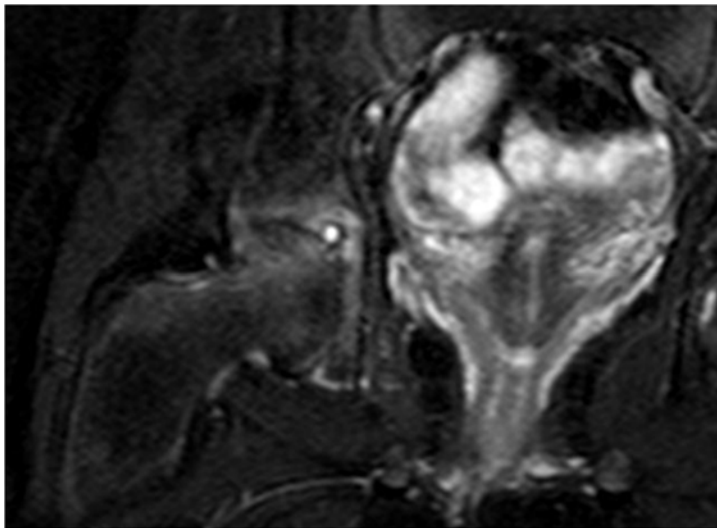


Figure 3: Presentative images from our series to demonstrate typical MRI features



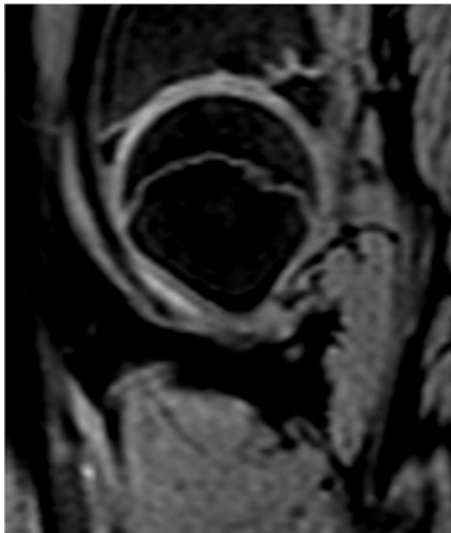
A) Coronal T2-weighted sequence showing JSN and oedema of the right hip.



B) Coronal T2-weighted sequence with fat suppression showing JSN and subchondral erosion on the acetabular side of the right hip.

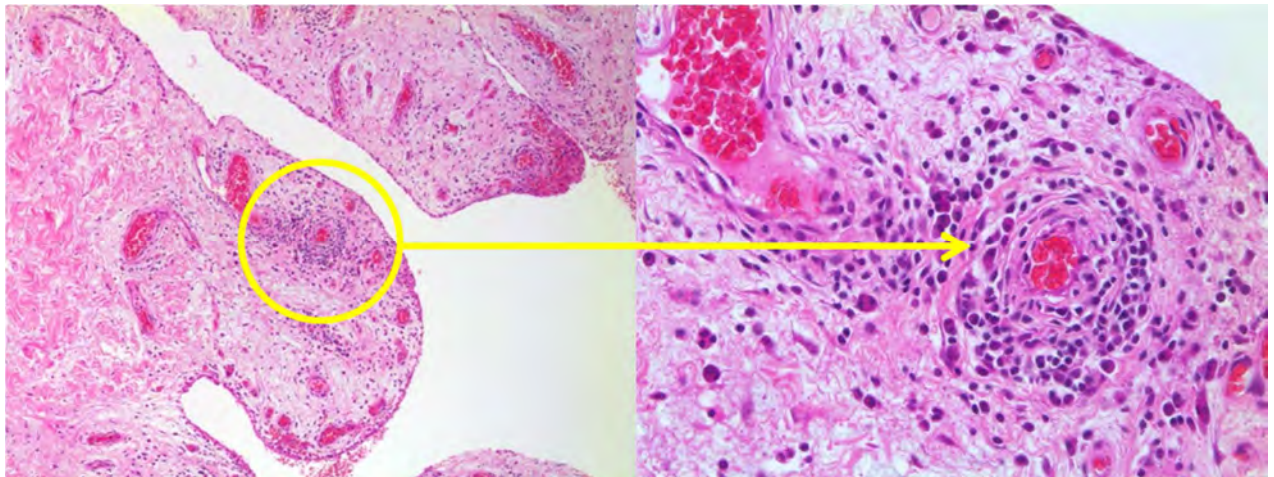


C) Coronal gradient echo sequence showing normal acetabular and femoral cartilage.

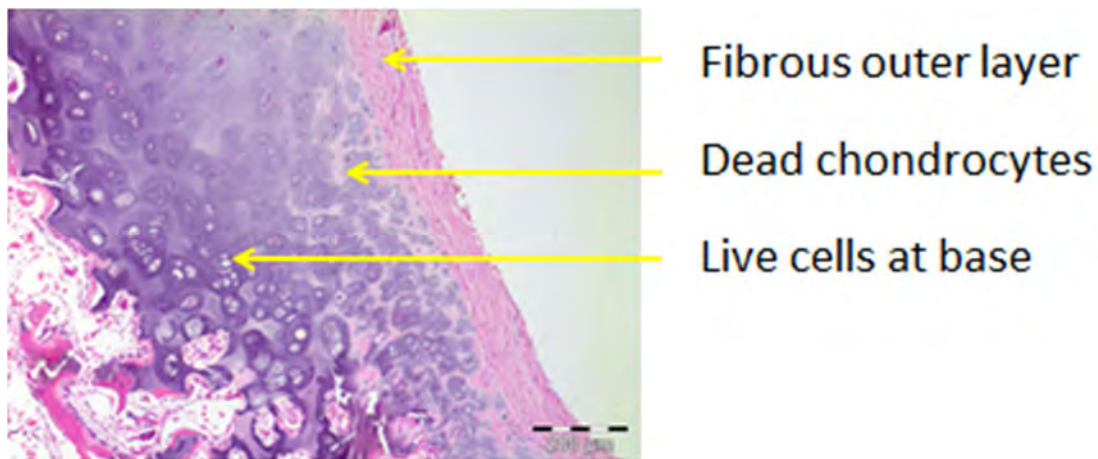


D) Sagittal gradient echo sequence showing acetabular cartilage loss.

Figure 4: Histological findings



A) Synovial biopsy showing a perivascular infiltrate of lymphocytes and plasma cells.



B) Cartilage biopsy showing an outer layer of fibroblastic tissue with an adjacent layer of degenerate chondrocytes and viable chondrocytes at the base.

PART D:

SUPPORTING DOCUMENTS

ETHICS APPROVAL LETTER



UNIVERSITY OF CAPE TOWN
Faculty of Health Sciences
Human Research Ethics Committee



Room E52-24 Old Main Building
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23 December 2013

HREC REF: 028/2013

Dr M Laubscher

Orthopaedics

H49

OMB

Dear Dr Laubscher

PROJECT TITLE: SUBTOTAL CAPSULECTOMY FOR IDIOPATHIC CHONDROLYSIS OF THE HIP

Thank you for submitting your study to the Faculty of Health Sciences Human Research Ethics Committee for review.

It is a pleasure to inform you that the HREC has formally approved the above-mentioned study.

Approval is granted for one year until the 30 December 2014.

Please submit a progress form, using the standardised Annual Report Form if the study continues beyond the approval period. Please submit a Standard Closure form if the study is completed within the approval period. (Forms can be found on our website: www.health.uct.ac.za/research/humanethics/forms)

Please note that the ongoing ethical conduct of the study remains the responsibility of the principal investigator.

~~PLEASE NOTE THAT THE ONGOING ETHICAL CONDUCT OF THE STUDY REMAINS THE RESPONSIBILITY OF THE PRINCIPAL INVESTIGATOR.~~

Please quote the HREC reference no in all your correspondence.

Yours sincerely

PROFESSOR M BLOCKMAN
CHAIRPERSON, FHS HUMAN ETHICS

Federal Wide Assurance Number: FWA00001637.

Institutional Review Board (IRB) number: IRB00001938

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	Therapeutic Studies— Investigating the Results of Treatment	Prognostic Studies— Investigating the Effect of a Patient Characteristic on the Outcome of Disease	Diagnostic Studies— Investigating a Diagnostic Test	Economic and Decision Analyses— Developing an Economic or Decision Model
Level I	<ul style="list-style-type: none"> High-quality randomized controlled trial with statistically significant difference or no statistically significant difference but narrow confidence intervals Systematic review² of Level-I randomized controlled trials (and study results were homogeneous³) 	<ul style="list-style-type: none"> High-quality prospective study⁴ (all patients were enrolled at the same point in their disease with ≥80% follow-up of enrolled patients) Systematic review² of Level-I studies 	<ul style="list-style-type: none"> Testing of previously developed diagnostic criteria in series of consecutive patients (with universally applied reference "gold" standard) Systematic review² of Level-I studies 	<ul style="list-style-type: none"> Sensible costs and alternatives; values obtained from many studies; multiway sensitivity analyses Systematic review² of Level-I studies
Level II	<ul style="list-style-type: none"> Lesser-quality randomized controlled trial (e.g., <80% follow-up, no blinding, or improper randomization) Prospective⁴ comparative study⁵ Systematic review² of Level-II studies or Level-I studies with inconsistent results 	<ul style="list-style-type: none"> Retrospective⁶ study Untreated controls from a randomized controlled trial Lesser-quality prospective study (e.g., patients enrolled at different points in their disease or <80% follow-up) Systematic review² of Level-II studies 	<ul style="list-style-type: none"> Development of diagnostic criteria on basis of consecutive patients (with universally applied reference "gold" standard) Systematic review² of Level-II studies 	<ul style="list-style-type: none"> Sensible costs and alternatives; values obtained from limited studies; multiway sensitivity analyses Systematic review² of Level-II studies
Level III	<ul style="list-style-type: none"> Case-control study⁷ Retrospective⁶ comparative study⁵ Systematic review² of Level-III studies 	<ul style="list-style-type: none"> Case-control study⁷ 	<ul style="list-style-type: none"> Study of nonconsecutive patients (without consistently applied reference "gold" standard) Systematic review² of Level-III studies 	<ul style="list-style-type: none"> Analyses based on limited alternatives and costs; poor estimates Systematic review² of Level-III studies
Level IV	Case series ⁸	Case series	<ul style="list-style-type: none"> Case-control study Poor reference standard 	<ul style="list-style-type: none"> No sensitivity analyses
Level V	Expert opinion	Expert opinion	Expert opinion	Expert opinion
<ol style="list-style-type: none"> A complete assessment of the quality of individual studies requires critical appraisal of all aspects of the study design. A combination of results from two or more prior studies. Studies provided consistent results. 				

4. Study was started before the first patient enrolled.
5. Patients treated one way (e.g., with cemented hip arthroplasty) compared with patients treated another way (e.g., with cementless hip arthroplasty) at the same institution.
6. Study was started after the first patient enrolled.
7. Patients identified for the study on the basis of their outcome (e.g., failed total hip arthroplasty), called "cases," are compared with those who did not have the outcome (e.g., had a successful total hip arthroplasty), called "controls."
8. Patients treated one way with no comparison group of patients treated another way.

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Journal article

1. Rand NS, Dawson JM, Juliao SF, et al. In vivo macrophage recruitment by murine intervertebral disc cells. *J Spinal Disord*. 2001; 14: 339-342.

Book chapter

2. Todd VR. Visual information analysis: frame of reference for visual perception. In: Kramer P, Hinojosa J, eds. *Frames of Reference for Pediatric Occupational Therapy*. Philadelphia, PA: Lippincott Williams & Wilkins; 1999:205-256.

Entire book

3. Kellman RM, Marentette LJ. *Atlas of Craniomaxillofacial Fixation*. Philadelphia, PA: Lippincott Williams & Wilkins; 1999.

Software

4. *Epi Info* [computer program]. Version 6. Atlanta, GA: Centers for Disease Control and Prevention; 1994.

Online journals

5. Friedman SA. Preeclampsia: a review of the role of prostaglandins. *Obstet Gynecol* [serial online]. January 1988;71:22-37. Available from: BRS Information Technologies, McLean, VA. Accessed December 15, 1990.

Database

6. CANCERNET-PDQ [database online]. Bethesda, MD: National Cancer Institute; 1996. Updated March 29, 1996.

World Wide Web

7. Gostin LO. Drug use and HIV/AIDS [*JAMA HIV/AIDS Web site*]. June 1, 1996. Available at: <http://www.ama-assn.org/special/hiv/ethics>. Accessed June 26, 1997.

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